HHS Public Access

Author manuscript

Ann Thyroid Res. Author manuscript; available in PMC 2017 September 01.

Published in final edited form as:

Ann Thyroid Res. 2017 ; 3(1): 95–101.

Primary Care Provider Management of Congenital Hypothyroidism Identified Through Newborn Screening

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Abstract

Objective—To assess Primary Congenital Hypothyroidism (CH) management patterns and feasibility of providing long-term care for patients with CH identified through newborn screening by Primary Care Providers (PCPs) in California and Hawaii.

Study Design—A survey was mailed to all physicians (N=823) listed as the referral doctor for confirmed patients with CH identified through newborn screening programs in both states between 01/01/2009–12/31/2013. Information was collected on CH management patterns, barriers to providing care, and knowledge on CH treatment. Descriptive statistics and bivariate logistic regression results were reported.

Results—206 PCPs completed the survey. Among these, 78% currently have patients with CH and 91% indicated willingness to provide long-term care to new patients with CH. Among PCPs currently caring for patients with CH, 17% managed CH by themselves with limited assistance from endocrinologists; 63% were involved in managing CH but endocrinologists played a larger role than PCPs; 19% were not involved in CH care. Only 49% of PCPs correctly answered questions regarding recommended follow-up frequencies and 23% knew the correct age for a trial off levothyroxine for suspected transient CH. Top two perceived barriers to providing long-term care included "need guidance or support from endocrinologists" (61%) and "not familiar with CH treatment guidelines" (28%).

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Conclusion—The majority of PCPs surveyed are willing to provide long-term care to patients with CH, but need support from endocrinologists and increased knowledge about current treatment guidelines.

Keywords

Congenital hypothyroidism; Primary care; Management; Newborn screening

Introduction

Primary Congenital Hypothyroidism (CH) is the most common preventable cause of neurocognitive disability (mental retardation) [1] and is the most common disorder diagnosed through Newborn Screening (NBS) [2]. CH affects about 1 in 2000 California newborns and 1 in 3000 Hawaii newborns, with over 245 new cases diagnosed in both states each year [2,3] CH refers to thyroid hormone deficiency due to dysfunction of the thyroid gland. About 80%–85% of permanent CH cases are caused by developmental failure or maldevelopment of the thyroid gland (*i.e.,* aplasia, hypoplasia, or ectopia). Other permanent cases are caused by an inherited enzyme deficiency affecting synthesis of thyroid hormone (dyshormonogenesis) or a thyrotropin receptor defect [1,4]. Although early detection and initiation of treatment within the newborn period have nearly eradicated severe mental disabilities among patients with CH in countries with NBS, unfavorable clinical outcomes are still present in a subset of patients [5]. CH severity and treatment inadequacy have been associated with a lower Intellectual Quotient (IQ) [6–9], lower educational attainment [10–12], and sensorineural hearing loss [10,13,14]. Patients with severe or inadequately treated CH also have increased risk of congenital malformations (e.g., anomalies of cardiac and nervous system and eyes), obesity [10,15–18], and impaired growth, puberty, and fecundity [19–22]. These findings highlight the importance of adequate treatment and monitoring to maintain optimal thyroid function throughout life to ensure the best neuro developmental, physical, and social outcomes in patients with CH.

Specific diagnostic, treatment, and follow-up guidelines for children with CH have been published by both the American Academy of Pediatrics (AAP) and the European Society for Paediatric Endocrinology [23,24]. Both guidelines recommend frequent follow-ups during childhood. However, a U.S. study using both public and private health insurance data found that over one-third of patients with CH discontinued treatment by three years of age [25]. Although the precise incidence of transient hypothyroidism is unknown (recent estimates range from 13.5%to 54.5% [26–28]), the authors of the analysis suggested that most cases of treatment discontinuation were likely not transient, but parent-initiated without physician supervision [23,25,27]. A follow-up study of patients with CH detected by the Michigan NBS Program showed that 25% of the patients were no longer being treated by three years of age, among which 83% had stopped treatment without medical supervision [29]. Of those who underwent a medically-supervised trial off medication, treatment was resumed in 87% of the cases [29]. These data suggest that discontinuation of treatment without medical supervision is a problem that could result in suboptimal outcomes for many patients. Due to the paucity of long-term follow-up data from the United States, it is unclear how patients

with CH are being managed as they age and the potential adverse effects of inadequate treatment or premature discontinuation of levothyroxine treatment.

In both California and Hawaii, patients who screen positive for CH in state NBS programs are usually referred by their Primary Care Providers (PCPs) to endocrinologists for further testing and confirmatory diagnosis. In California, a large percent of screen positive patients are referred to state-contracted endocrine centers to provide follow-up care. However, feedback from endocrinologists indicated that these centers usually do not have the resources needed to follow every patient with CH as frequently as recommended by the AAP guidelines, especially during the first three years of life, when treatment adherence is most critical. Furthermore, it is often more practical and convenient for patients with CH to receive care from their PCPs who are usually in closer proximity and more readily available, especially in rural areas where fewer endocrinologists are available [30]. Parks et al found that in Texas during 2004–2006, 64% of patients with CH were followed and reported by endocrinologists in the first year of life and the proportion decreased to 26% in the second year of life. Underreporting from endocrinologists might exist, but these findings indicated that some patients might be followed by primary care pediatricians [31]. The AAP-recommended CH follow-up visits can be completed during routine well-baby visits. Endocrinologists can serve as consultants after the initial evaluation of the case is completed and provide guidance about modifying treatment when questions arise. The consensus is that PCPs should take more responsibility for providing long-term care and encouraging adherence to treatment for patients with CH with the support and consultation from pediatric endocrinologists.

These recommendations are consistent with the patient-centered medical home model for providing primary care that has been widely endorsed by physicians' associations, including the AAP and the American Academy of Family Physicians [32]. Studies have shown that, for children and youth with special health-care needs, care provided through a medical home is associated with better care coordination, higher satisfaction with care, and fewer emergency room visits or hospitalizations [33–36]. Compared to metabolic disorders screened for by NBS, CH is easier to manage since it primarily involves the administration of a single drug. Thus, with proper training, the locus of the medical home can be with the PCP when that provider is supported by apediatric endocrinologist for challenging and more nuanced cases. Increased PCP involvement and "ownership" of care for patients with CH would likely improve quality of care, disease management and health outcomes. Understanding the hurdles impeding management of children with CH by pediatricians and whether it is feasible to shift the Long-Term Follow-Up (LTFU) responsibility to PCPs with support from pediatric endocrinologists will strengthen the medical home for these patients.

The purpose of this study was four-fold: 1) to evaluate the current CH case management patterns; 2) to assess the willingness and capability of PCPs to provide LTFU for patients with CH; 3) to identify potential barriers for PCPs to provide LTFU to patients with CH; and 4) to assess PCPs' willingness to obtain informed consent and provide data to an existing database to evaluate quality of care and patient outcomes for possible research endeavors in the future.

Methods

A cross-sectional survey of PCPs was conducted from February through June of 2014. All PCPs who were listed as the referral doctors for at least one patient with CH born between 2009–2013 were selected from the California and Hawaii NBS databases. A total of 801 physicians from California and 22 physicians from Hawaii were invited to participate in the study.

The survey included five key components relevant to PCPs: 1) current practice in managing patients with CH; 2) barriers and resources needed to conduct CH LTFU; 3) willingness and capability to conduct CH LTFU; 4) willingness and capability to obtain informed consent from patients and provide data to a LTFU database; and 5) clinical outcomes of patients with CH being seen by the PCPs. Results of the fifth component will be reported in a future paper. To maximize response rate, we included a personalized cover letter in the survey-mailing packet that described the significance and objectives of the study. To better understand the reasons for non-response, we asked each doctor who was unable to complete the questionnaire to indicate reasons for non-response on a separate “Non-Response Card” enclosed with the initial mailing. A gift card incentive was offered to PCPs who completed either the full survey or the non-response card. An on-line version of the same survey was also made available at the same time. One hundred and thirteen mailing packets were returned as undeliverable after the initial mailing. We were able to obtain their most up-to-date addresses through multiple modes (*e.g.,* internet search and calling the listed offices) for 87 doctors and resent the packets. We failed to find usable addresses for 26 doctors and they were excluded from the response rate calculation. Additionally, we sent out two reminder letters to non-responding PCPs, one and two months after the initial mailing, to boost the response rate.

All data were analyzed using SAS version 9.3 for Windows (SAS Institute, Cary, NC). Continuous variables were expressed as medians with range and categorical variables were presented as numbers and percentages. Descriptive statistics were calculated and the differences between the two states were assessed using Chi-square and Fisher’s exact tests for categorical variables and Wilcoxon Signed Rank Test for continuous variables to compare the difference in medians. Bivariate logistic regression modeling was used to assess the association between selected covariates and key outcome variables. Crude Odds Ratios (ORs) and their 95% Confidence Intervals (CIs) were reported. This study was approved by the State of California, Health and Human Services Agency, Committee for the Protection of Human Subjects, Project Number 13-08-1317.

Results

Response rates and demographics

A total of 238 doctors in California and 10 doctors in Hawaii responded to the mailing, among whom 226 completed the survey (128 paper surveys and 98 on-line surveys) and 19 returned the Non-Response Card (Figure 1). Among doctors who completed the survey, 20 were not PCPs and their responses were excluded from the final analysis. The total response rate was 28% for California doctors and 45% for Hawaii doctors. Among the 19 doctors/

offices that returned the Non-Response Card and indicated a reason for not completing the survey, four did not have time to do the survey, seven were specialists instead of PCPs, six did not have patients with CH, one doctor was deceased, and one had security concerns.

Among the 206 PCPs who completed the survey, 51% were male, 46% were White, 39% were Asian, 7% were of Hispanic origins and 8% were of other race or ethnicity (Table 1). The majority of respondents were primary care pediatricians and 8% identified themselves as family physicians. Nearly 50% were in private practice, 33% were in group practice, 13% were in community health centers, and only 4% were in a hospital-based practice or HMO. Respondents had been in medical practice for a median of 18 years (range 2–43 years). There was no statistically significant difference in these variables between respondents from California and Hawaii.

Case management patterns and long-term care feasibility

When California PCPs were asked who usually manages CH for their patients, 17% indicated that their patients' CH was mainly managed by themselves, but endocrinologists were involved; 63% were involved, but patients' CH was mainly managed by endocrinologists; and 19% indicated that their patients' CH was solely managed by endocrinologists. Among Hawaii PCPs, 75% indicated that their patients' CH was mainly managed by endocrinologists, but they were also involved and the rest indicated that their patients' CH was solely managed by endocrinologists. Among all respondents, "need guidance or support from endocrinologists" and "not familiar with the CH treatment guidelines" were listed as the two most commonly perceived barriers for PCPs to provide long-term care for patients with CH.

Over 90% of responding PCPs in both states indicated willingness to provide long-term care for new patients with CH. Compared to PCPs who mainly manage patients' CH by themselves, those who were not involved in their patients' CH management had 85% lower odds of being willing to provide long-term care for new patients with CH (OR=0.15, 95% CI: 0.03, 0.77). Type of practice and currently having patients with CH were not associated with willingness to provide long-term care for new patients with CH (Table 2 and 4).

Over two-thirds of PCPs in both states expressed willingness to obtain informed consent from patients to record and share LTFU data on their patients with CH. Nearly 75% of PCPs consider obtaining informed consent doable with little or no difficulties. About 87% of PCPs from both states indicated willingness to report LTFU data if given a reasonable compensation. PCPs currently caring for patients with CH were more likely to indicate willingness to provide LTFU data than those who had no patient. Among the 27 California PCPs who were not willing to collect LTFU data, over one-third indicated "do not have enough staff or time to enter data" or "do not provide long-term care for patients' CH condition" as the main reasons for their unwillingness to do so. In terms of compensation required to provide LTFU data, 73% of PCPs would be satisfied with a compensation less than $200 per patient per year (Tables 2 and 4).

Knowledge about CH-related management

When asked about the recommended frequency of blood tests for patients with CH in three different age groups (<6 months, 6 months to 3 years, and >3 years), the proportion of PCPs who correctly answered each question was72%, 60%, and 73% for each age group, respectively. Only 49% of PCPs correctly answered all three questions about the recommended follow-up frequencies (Table 3). There was no statistical difference in the prevalence of knowing the recommended follow-up frequencies between PCPs who reported different management patterns, type of practice, or number of patients (Table 4).

When assessing PCPs' familiarity with indications for a trial off levothyroxine therapy to assess the transient status of CH, only 2% were very familiar, 28% were somewhat familiar, 55% were not familiar, and 14% did not know. Only 23% of PCPs in both states knew the recommended age for such a trial (Table 3).

Preference for CH-related continuing medical education

Over 80% of responding PCPs in both states indicated being "likely" or "very likely" to participate in Continuing Medical Education (CME) about CH if such courses are available. PCPs who reported currently having patients with CH were more likely to participate in CH-related CME than those without patients (Table 4). With regard to the preferred format for CME, over 60% of respondents chose webinars, about one-third chose in-person classes, and one-third chose grand rounds presentations.

Discussion

This study of current management patterns of patients with CH and feasibility of PCP involvement in long-term care provides important insights on how to assure high quality primary care and optimal clinical outcomes for patients with CH. Although the majority of PCPs who completed the survey were willing to provide long-term care to their patients with CH, endocrinologists were identified as assuming the primary management responsibilities in most of the cases in both states. The two most commonly perceived barriers by PCPs to providing long-term care for patients with CH were: needing guidance from endocrinologists and lack of familiarity with the current CH treatment guidelines. We identified a general lack of knowledge about CH treatment and management among responding PCPs. Nonetheless, the majority of respondents were willing to provide long-term care for new patients with CH. Specifically, they considered it feasible to obtain informed consent from patients with CH or their guardians, and would be willing to provide LTFU data to an existing database for a compensation of $200 or less per patient per year.

Long-term follow-up data for patients identified through NBS can help evaluate the clinical outcomes and health service utilization patterns of patients with diagnosed disorders in diverse populations. Multiple national agencies have released initiatives to establish a comprehensive, sustainable, and feasible long-term follow-up data collection system to improve the ultimate health outcomes of patients with screened disorders [37–40]. For patients with CH, optimal treatment throughout life is critical for achieving the best neurological, physical, economic, and social outcomes [24,40]. How to effectively deliver

the needed care to all patients with CH and ensure adequate treatment for everyone remains a challenge in both California and Hawaii. National data showed that there is a shortage of pediatric endocrinologists nationwide; the average time that patients have to wait to see a pediatric endocrinologist is 10.3 weeks and the average distance to pediatric endocrinology care is over 26 miles [30]. Findings from this study indicate the possibility of integrating such care into primary care practices. The majority of PCPs in both states were willing to take the responsibility of caring for patients with CH with support and guidance from endocrinologists. Future efforts should be focused on how to improve the communication between PCPs and endocrinologists and to ensure that each patient receives appropriate and comprehensive care.

To our knowledge, this is the first survey of PCPs to identify a knowledge gap in CH management. To increase PCPs' capability for providing long-term care for patients with CH, one avenue would be to provide CME opportunities for PCPs to learn about current guidelines for CH treatment and management. In response to the documented strong interests in CME training from responding PCPs, we have since developed a CH management curriculum entitled, "Congenital Hypothyroidism: What Every Pediatrician Needs to Know." This course has been offered through Pediatric Grand Rounds throughout California and Hawaii. Building off of this CME course for PCH providers, Stanford University recently developed an online CME course on CH that is applicable more broadly to pediatric care providers (https://med.stanford.edu/cme/courses/online/hypothyroidism.html). These teaching modules review how to confirm diagnosis, initiate treatment if required, monitor thyroid function, adjust levothyroxine dosing for infants, children and adolescents, and underscore the need for providers to educate families about the importance of adherence to treatment. We hope that such efforts will increase the confidence and competence of PCPs to manage patients with CH and improve their health outcomes.

We recognize a key limitation in this study. The overall response rate of completed surveys was 28.4%, despite the substantial efforts to improve the response rate as outlined in methods section [41]. The top three reported reasons for not completing the survey among those who returned the Non-Response Card were: being specialists themselves, having no time, and not currently having any patient with CH. The first two reasons may not cause any response bias. The third reason, not currently caring for any patient with CH, could result in an overestimation of several outcome variables including the percentage of doctors currently having patients with CH and knowledge about CH management. It is possible that PCPs currently caring for patients with CH would be more likely to participate in the study and have better knowledge than those without any patient with CH. Prior studies have shown that provider surveys are resistant to non-response bias and our response rate was better than other provider surveys of similar length [41–44]. We believe that the findings may be reasonably representative of PCPs in both states.

Conclusion

PCPs in both California and Hawaii expressed willingness to provide long-term care for patients with CH, but to fulfill the responsibility they need additional training on up-to-date CH-related treatment and management guidelines, as well as strong support from

endocrinologists who can provide consultation for dealing with difficult cases. In addition, PCPs stated willingness to provide long-term follow-up data for patients with CH to existing databases. In future efforts to improve the clinical outcomes for patients with CH or other disorders identified by NBS and ensure that they receive high quality primary care services we should try to better engage PCPs in the care process and improve the collaboration between PCPs and specialists. More research is needed to assess the effectiveness of educational efforts for PCPs and the feasibility of conducting long-term follow-up for rare diseases in primary care settings.

Acknowledgments

We would like to thank the Human Resources and Services Administration for providing funding for this study. We also thank all doctors who participated in the survey, and colleagues at the California Genetic Disease Screening Laboratories and Genetic Disease Screening Program, including, Tracey Bishop, Shellye Lessing, Norah Ojeda, Bob Currier, Sona Saha and Hari Haran,for their help with the survey contents and administration. Drs. Ning Rosenthal and Lisa Feuchtbaum had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. This project was supported by the Health Resources and Services Administration (HRSA) of the U.S. Department of Health and Human Services (HHS) under grant D93MC26187-02-00: Demonstration Projects for Integrating Newborn Screening Long Term Follow-Up into Primary Care Practice. This information or content and conclusions are those of the authors and should not be construed as the official position or policy of, nor should any endorsements be inferred by HRSA, the Centers for Disease Control and Prevention, HHS or the U.S. Government. None of the coauthors has conflicts of interest relevant to this article to disclose.

Funding Source

This project is supported by the Health Resources and Services Administration (HRSA) of the U.S. Department of Health and Human Services (HHS) under grant D93MC26187-02-00: Demonstration Projects for Integrating Newborn Screening Long Term Follow-Up into Primary Care Practice. The funding agency does not have a conflict of interest with this research or this manuscript and did not influence (1) the study design; (2) the collection, analysis, and interpretation of data; (3) the writing of the report; and (4) the decision to submit the paper for publication.

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Abbreviations

CH	Primary Congenital Hypothyroidism
NBS	Newborn Screening
LTFU	Long-Term Care and Follow-Up
PCP	Primary Care Providers
AAP	American Academy of Pediatrics
CME	Continuing Medical Education

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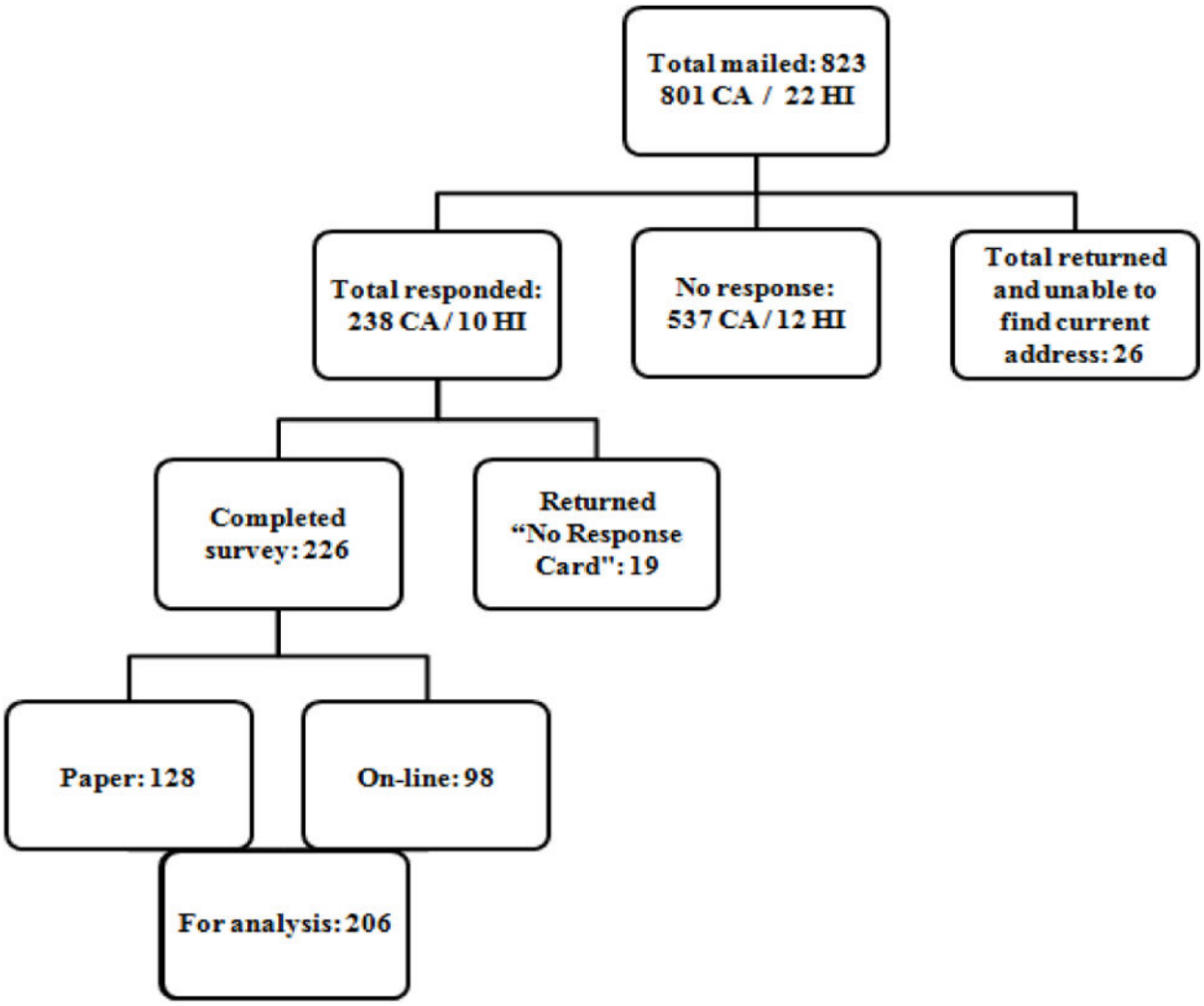

Figure 1.
Response rate of the Primary Care Providers (PCPs) surveyed in California (CA) and Hawaii (HI) in 2014.

Table 1

Characteristics of the responding primary care providers in California (CA) and Hawaii (HI).

Characteristics	CA (%, n=196)	HI (%, n=10)	All (%, n=206)
Male sex	51	60	51
Race/ethnicity			
Hispanic/Latino	7	0	7
White	47	20	46
Asian	38	60	39
Black	3	0	3
Mixed or Other	5	20	5
Medical specialty			
Family practice	9	0	8
Pediatrics	91	100	92
Type of practice			
Private practice	45	50	45
Group practice	33	30	33
Hospital-based practice	5	0	4
HMO	4	0	4
Community health center	13	20	13
Years in medical practice			
Median (range)	18 (2, 43)	19 (8, 29)	18 (2, 43)

Note: There was no statistically significant difference between CA and HI for any of the above variables.

Table 2

Case management patterns, perceived barriers, and willingness to providing Long-Term care and Follow-Up (LTFU) for patients with primary Congenital Hypothyroidism (CH).

Variables	CA (%)	HI (%)	All (%)
Who manages patients' CH conditions			
Solely by PCPs	1	0	1
Mainly by PCPs, but endocrinologists are involved	17	0	16
Mainly by endocrinologists, but PCPs are involved	63	75	64
Solely by endocrinologists	19	25	19
Willing to provide long-term care for new patients with CH			
Yes	66	80	67
Maybe	25	20	24
No	6	0	6
Don't know	3	0	3
Perceived barriers for providing long-term care			
Need guidance or support from endocrinologists	60	80	61
Not familiar with the CH treatment guidelines	28	40	28
Need more staff time to coordinate care	15	10	15
Patients are not compliant with care	14	10	14
Don't have enough time	12	10	12
CH long-term care is too complicated	11	0	10
Don't get enough reimbursement	11	0	10
Anticipate no barriers	20	10	20
Willingness to obtain informed consent	75	100	76
Difficulties in obtaining informed consent			
Very difficult	4	0	4
Difficult, but doable	35	20	34
Not difficult at all	39	70	40
Don't know	22	10	21
Willingness to provide LTFU data			
Yes	67	90	68
Maybe	19	10	19
No	3	0	3
Don't know	11	0	10
Reasons for not willing to provide LTFU data			
Don't have enough staff or time to enter data	37	0	37
Do not provide long-term care for patients' CH	37	0	37
Have concerns over patient's privacy	19	0	19
The LTFU data is not relevant to clinical practice	7	0	7
It is not important to collect LTFU data for CH	0	0	0

Variables	CA (%)	HI (%)	All (%)
Compensation required to provide LTFU data			
$50/patient/year	5	20	6
$100/patient/year	18	20	18
$150/patient/year	14	0	13
$200/patient/year	18	0	17
No compensation is needed	18	40	19
Don’t know/Other	27	20	27

Note: There was no statistically significant difference between California and Hawaii for any of the above variables. CA: California; HI: Hawaii.

Table 3

Knowledge of current treatment guidelines for primary congenital hypothyroidism.

Variables	CA (%)	HI (%)	All (%)
Know the recommended frequency of blood tests			
In the first six months of age	72	70	72
Between six months and three years of age	59	70	60
Over three years of age	74	50	73
Know all frequencies	49	50	49
Familiarity with indications for trial off levothyroxine			
Very familiar	3	0	2
Somewhat familiar	29	20	28
Not familiar	54	70	55
Do not know	15	10	14
Know at what age for trial off levothyroxine	23	30	23

Note: There was no statistically significant difference between California and Hawaii for any of the above variables. CA: California; HI: Hawaii.

Table 4

Association between selected covariates and key outcome variables.

Covariates	Know recommended follow-up frequencies OR (95% CI)*	Willing to provide care for new patients with CH^ (Yes and Maybe) OR (95% CI)*	Willing to provide LTFU data (Yes and Maybe) OR (95% CI)*	Likelihood to participate in CME (Very likely and Likely) OR (95% CI)*
Currently having ≥1 patients with CH^				
Yes	1.1 (0.5, 2.1)	1.8 (0.6, 5.4)	3.1 (1.3, 7.6)	3.0 (1.1, 8.5)
No	Reference	Reference	Reference	Reference
Management patterns				
Mainly by PCP$, but endocrinologists are involved	Reference	Reference	Reference	Reference
Mainly by endocrinologists, but PCP is involved	0.9 (0.4, 2.0)	4.1 (0.5, 30.6)	1.3 (0.3, 5.2)	3.0 (0.8, 11.4)
Solely by endocrinologists	1.2 (0.4, 3.2)	0.15 (0.03, 0.77)	0.6 (0.1, 3.0)	1.2 (0.3, 5.3)
Type of practice				
Private practice	Reference	Reference	Reference	Reference
Group practice	1.1 (0.6, 2.0)	1.1 (0.4, 3.0)	1.6 (0.5, 4.8)	0.4 (0.1, 1.2)
Hospital-based practice	0.12 (0.01, 1.04)	1.0 (0.1, 8.7)	0.25 (0.05, 1.14)	0.22 (0.04, 1.35)
HMO$	1.7 (0.4, 7.4)	N/A	0.12 (0.03, 0.57)	0.22 (0.04, 1.35)
Community health center	1.7 (0.7, 4.1)	3.2 (0.4, 26.3)	0.5 (0.2, 1.7)	1.8 (0.2, 15.5)

*OR: Odds Ratio; CI: Confidence Interval;

^CH: Primary Congenital Hypothyroidism

Significant odds ratios are shown in bold.

$PCP: Primary Care Providers; HMO: Health Maintenance Organization